

RESEARCH ETHICS

It doesn't cost anything just to ask, does it? The ethics of questionnaire-based research

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Patient-based outcome measures are increasingly important in health care evaluations, often through the use of paper-based questionnaires. The likely impact of questionnaires upon patients is not often considered and therefore, the balance of benefit and harm not fully explored. Harms that might accrue for research staff are even less frequently considered. This paper describes the use of postal questionnaires within a study of breast disease management in primary care. Questionnaire responses are used to describe the nature of discomfort or harms that may occur in such studies. Ethical issues raised by the harms are discussed in relation to the benefits of the study. Practical suggestions for reducing harm to patients are proposed. A secondary consideration, discomfort to the researcher, is also identified and suggestions made to reduce its effect. Finally, the role of research questionnaires as a study intervention is discussed.

Traditionally, health care interventions have been assessed using predominantly biomedical outcome measures. Increasingly though, the limitations of this approach have been recognised with a commensurate rise in patient-based outcome measures, often involving questionnaires.¹ It might be supposed that, in considering the ethics of medical research, the most important and weighty ethical issues would arise in experimental studies involving clinical interventions such as drugs or surgery, rather than in observational or questionnaire-based studies. This supposition needs scrutiny.² Inherent in it is an assumption that the means of collecting data—that is, making observations and asking questions—do not amount to interventions and therefore have no impact upon patients. We shall present and discuss a single case of a questionnaire-based study, which shows how real and active an intervention the simple asking of questions can become. Furthermore, the potential risks of questionnaire-based methods in general and how they may be reduced will be discussed.

It has been argued that the dominance of the medical paradigm in the bioethics literature has resulted in a greater focus upon the experimental method compared to different methodological approaches.³ Middle *et al* reported their experience of ethical submissions to 162 local research ethics committees (LRECs) for a survey of parents of National Health Service patients.⁴ They revealed that 10% of LRECs considered it inappropriate to apply for ethical approval for such a study. The view that epidemiological surveys are “safe” and “non-invasive” has been clearly proposed.⁵ A distinction between intrusive and non-intrusive research is useful here.⁶ Intrusive research involves direct involvement with patients and may be either invasive or non-invasive. Either way, the ethical issues should be considered more carefully, for intrusive research and questionnaire studies fall within this category.

A recent review argued that participants in qualitative research may risk significant harm (for example, emotional wellbeing).⁷ One evaluation of the impact on women of being interviewed about the aetiology of cervical cancer, however, found that half the sample identified benefits from participation and only two women (<1%) regretted taking part.⁸ Stress was considered unlikely to be caused when interviews were conducted by experienced interviewers. More recently, re-

spondents to a postal survey of mental health reported few problems with distress caused by their participation and many reported feeling better.⁹

A distinction should be made between qualitative approaches (for example in-depth interviewing), and structured postal questionnaires. The ability to interact with research participants differs greatly between the two methods, with significant flexibility in the interview setting.³ Questionnaires using standardised measurement scales are increasingly common in health services research, decreasing the opportunity to respond directly to patients.

In all clinical research, the key ethical issues are primarily whether the research is justifiable in terms of the balance of possible benefits and harms for the research subject and, if it is justifiable, whether the subjects properly consent to participation.² However, a secondary, and less well-recognised, set of issues concerns the possibility of harm to participating professionals.¹⁰ Ethical review of clinical research should consider these issues as well. The following case illustrates both these primary and secondary moral concerns.

THE QUESTIONNAIRE

The BRIDGE study is a randomised trial of the implementation of guidelines for women presenting with breast disorders in primary care.¹¹ The aim of the guidelines is to reduce referral of women with benign symptoms and with no increased risk of cancer: referrals may be driven in part by women's fear of cancer. Since these referrals may lead to unnecessary biopsies—potentially causing physical and psychological morbidity—the importance of successful and appropriate implementation of the guidelines seems self evident. Hence the value of a questionnaire, which establishes and clarifies patients' views on their management, also seems necessary. A secondary aim, of evaluating women's perception of risk from breast symptoms, was also considered necessary to assess the need for risk-communication strategies. It is this study of management expectation and risk-perception that the questionnaire and this paper examines. Approval for the study was granted by the relevant LRECs.

A large number of women were to be recruited to the study ($n > 1000$) and were likely to present with a variety of clinical symptoms and represent similarly varied personal experiences

(cases). The survey was also to include women who had not recently presented to their general practitioner (GP) and who may never have experienced breast problems (controls). To capture this variety for a representative cross-section of women, a quantitative questionnaire approach was chosen. Whilst predominantly comprised of closed questions offering fixed response categories, space was provided in the questionnaire for respondents to provide free-text (their own) comments. Two examples of the fixed response questionnaire items are as follows:

In your opinion is a woman who has had painful breasts more likely to suffer from breast cancer sometime?

If you had painful breasts how important would it be for you that your GP sent you to the hospital breast clinic/outpatient clinic?

Women presenting with a breast complaint were approached for written consent by their GPs. Age-matched control patients were selected from practice registers. Lists of women selected as controls were reviewed by their GP to ensure that vulnerable patients were not approached. Controls were sent an introductory letter and a copy of the postal questionnaire. Both cases and controls received the same version of the questionnaire. Patients' agreement was sought for the collection of clinical data about their presentation (cases only), and to the completion of a confidential postal questionnaire about their experience of breast disease, their expectations for management, their knowledge about breast cancer (for example epidemiologic and aetiological factors), and their perceptions of their own personal risk of developing various breast diseases (including cancer). All patients were provided with a study information sheet and provided written consent.

Nine hundred and fifty-nine questionnaires were returned (497 cases/462 controls), a response rate of 85%. One third ($n=318$) of respondents made free-text comments. Most comments described experiences of general practice and outpatient clinic management.¹² Many women reported satisfaction with their management, whilst a few respondents used the questionnaire as an opportunity to raise discontent, express worries, and seek reassurance and advice.

RESPONSES CAUSING CONCERN

A small number of respondents ($n=29$) made negative comments about care received in primary care. Mostly, these comments did not suggest mismanagement and often related to the care of women other than those completing the questionnaire itself (for example friends). However, three specific responses following the questionnaire particularly concerned the researchers:

- 1 One woman detailed her family history of breast cancer and asked what her chances were of getting breast cancer.
- 2 Another woman on hormone replacement therapy, anxious about possible risks from her treatment with regard to breast cancer, wanted to know if she should continue taking her treatment.
- 3 A third woman telephoned the researcher on receiving the questionnaire. She had presented to her GP four weeks previously and was angry at receiving the questionnaire so quickly whilst still waiting for an outpatient clinic appointment.

These responses are similar to those encountered in other similar questionnaire studies.¹³

THE PROBLEMS THESE RESPONSES PRODUCED

The key considerations seem to arise from judging the researcher's own role and responsibilities in regard to this

questionnaire. The aim of the research was to improve health care generally, and not to intervene in the management of particular cases. The first two women were asking for clinical advice following their consultation and like many people they took the opportunity to seek other opinions. Giving clinical advice is inappropriate for researchers with no clinical responsibility for the patients in question. Furthermore, for the researcher to attribute clinical risk would be impossible without further clinical contact with the patient.

A more extreme concern is that patients may describe care which has been in some way deficient. The observation of inadequate care whilst undertaking research has been discussed previously, although not in the context of postal questionnaires.¹⁴

Somewhat parallel considerations arise in the case of the third woman, who was expressing her dissatisfaction with important administrative aspects of her management. Again it is clear that the researcher has no mandate or authority to act as the patient's advocate (for example by attempting to expedite referral). Given the limited information available it is difficult to determine whether mismanagement was likely. If the GP suspected breast cancer, then the delay in receiving an outpatient appointment would be clearly inappropriate.¹⁵ If, however, the patient was referred under other clinical circumstances, then the delay is more difficult to interpret without clinical details.

Finally, all three cases put the researchers in the position of unwilling and unauthorised auditor of clinicians whose identity was known to them from study organisation and data collection.

ETHICAL ISSUES (I): HARM TO THE PATIENTS

The recruited women were presenting with unspecified breast symptoms which might resolve into a variety of diagnoses, ranging from the trivial to the life-threatening. At the time of recruitment into the study they did not know their diagnosis. Even at the time the questionnaire was received (four weeks on) there was no guarantee that confirmed diagnoses would have been established. Thus there is a distinct risk that the initial invitation and, for a few women, the content of the questionnaire itself might create or reinforce anxiety about life-threatening illnesses in subjects who do not have such illnesses. It is true also that the same anxieties may be generated in subjects who do in fact have the corresponding illnesses. It is worth identifying this as a distinct harm because the anxiety in this case is not groundless and it may be even more unpleasant to deal with than that experienced by the former group.

These first two harms concern the raising of worry or anxiety. Such harms have been recognised previously, although seldom in the context of this type of research.² Whilst no evidence of this occurring in the present study was apparent, it is nevertheless a possibility in a study of this nature. Indeed, two other women in this study commented that "other" women might find the questions worrying, although they themselves did not. A third, and in this case, least foreseen possible harm is that, through containing the opportunity for free-text comment, the questionnaire will give rise to more positive expectations which, since they may not be met, will lead to subsequent disappointment: namely, expectations of obtaining wider information (level of risk associated with family history and other potential risk factors) or of obtaining further opinions or even assistance regarding current clinical management. The three examples of patient feedback above relate to this harm. These expectations arise where subjects misunderstand the role and responsibilities of the researchers. Not unreasonably, patients may look to the researcher for information and may be disappointed when the researcher is unable to comply.

Table 1 Patient harms

Harm identified	Reducible by:
1 Increased anxiety in patients with no pathology	Delay questionnaire until diagnosis clear
2 Increased anxiety in patients with pathology	Reminder/reassurance about withdrawal and leaving problematic questions
3 Unrealistic positive expectations	Adequate information at consent stage to clarify the role of the researcher; established methods for patient to re-contact the primary care team

If these are the possible harms, then two questions arise: first, are they avoidable and second, if unavoidable could they none the less in principle be justified by the possible benefits of the research? We may not need to ask this second question if the harms are straightforwardly avoidable, and it seems that two of them are. The first harm can be avoided by ensuring that no questionnaires are distributed to subjects who have not already received their clinical diagnosis. This may of course involve liaison between the researcher and the subjects' doctor, but seems a modest enough requirement, as does having to accept any minor delays. In other words, the design of the study could remove the needless or otherwise inappropriate anxiety, which the questionnaire might generate in women whose illness is not in fact serious or life-threatening. The third harm is avoidable (or at least reducible) by ensuring that the research subjects are given enough information, and in a sufficiently clear manner, at the time of giving consent so that there are no reasonable grounds for them to form misconceptions about the researchers' roles and responsibilities which give rise to unrealistic expectations. An alternative or, better, additional measure would be to identify a named clinician who could deal in a responsible way with any inquiries that the subjects might have with regard to information or advice about their current clinical management. Such responses would of course require great care and an agreed policy of strict directing of specific concerns to the clinician with direct responsibility for a given patient. Some difficulties may remain, nevertheless, a named clinician with this general role would be better able to field and distribute inquiries because of his or her professional competence and status.

The second harm (increasing anxiety in women with serious pathology) is more difficult. It is hard to see how this harm can be avoided. Women who have received a diagnosis of serious or life-threatening illness will find questions about their expectations of treatment to be uncomfortable, and questions about their perceived risk of developing the disease to be sourly ironic. This suggests, perhaps, that these questions about risk perception ought to be reserved for women whose diagnosis rules out such illnesses at the present time. Alternatively, a reminder could be included at the start of each questionnaire that patients are not obliged to answer any question they find upsetting or inappropriate. Questionnaire design would, however, normally seek to engage the respondent as fully as possible in answering each item in order to enhance validity.¹⁶ There is a balance to be struck and the study design would need to be examined to assess the impact of such item non-completion. Furthermore, any ethically sound research must include a reminder to subjects that they may withdraw from the research at any time without giving a reason.

In all events, there is one further requirement. All subjects must enter the study with their eyes open. It would, in particular, need to be pointed out to women considering participation that they might be asked questions which might be especially uncomfortable, given that they would now, under the revised form of the study, complete the questionnaire when their diagnosis was known. Clearly subjects cannot fully envisage this situation at the time when they are first recruited (at an early clinical consultation). Nevertheless, they must be invited to contemplate how they might feel if informed consent is to be promoted.

The consent stage, therefore, is the place to face up to and deal with one of the three harms we have identified (provoking anxiety in those with serious disease), and also the place simply to avoid one of the others (the generation of unrealistic and inappropriate expectations). The remaining harm concerning provoking anxiety in those who do not have serious disease, can be designed out (see table 1).

ETHICAL ISSUES (II): HARM TO PARTICIPATING PROFESSIONALS

The primary issues should be regarded as those concerning the interests and wellbeing of the research subjects. However, the possibility of harms to the researcher is also ethically important and deserves consideration. In this case the harms to the researchers are concerned with the expectations some patients have of them. It is problematic for a researcher to be mistakenly identified as a source of help which he or she cannot give, particularly in view of the serious and distressing nature of the illnesses which are in the forefront of these patients' minds. It would not be surprising if this led to an understandable, if inappropriate, sense of guilt on the part of a researcher.

Coupled with this is the possibility that a participating GP in good faith provides opportunities for patient approaches and the collection of clinical data, only to find himself/herself on the receiving end of unexpected and unsolicited clinical audit.

Clearly these harms need to be foreseen and accounted for in the design of any future study of this kind. They strongly suggest that a training procedure be established for the participating professionals. Researchers will inevitably encounter difficult clinical issues in the course of their work. Preparatory work involving say, the use of role-play may help to raise the researchers' awareness of the potential problems and equip them with skills to manage such encounters. Most problems, however, will need to be taken back to the GP by the patient, and appropriate verbal and written materials can be prepared to facilitate this (which is what occurred in this study). Included explicitly within such preliminary work could be a consent procedure parallel to that established for the clinical subjects, so that researchers avoid finding themselves in an invidious, inappropriate, and disturbing situation without having faced the possibility beforehand, in an informed and reflective way.

WHEN IS AN INTERVENTION NOT AN INTERVENTION?

Ethical guidelines for researchers in medicine and nursing are well established and describe appropriate means of assessing risk, informing patients about research and obtaining consent.¹⁷⁻¹⁹ Within these guidelines, methods such as questionnaires are not specifically excluded, but decisions such as risk assessment are mostly expressed in relation to experimental trials. This may create an impression that data collection methods such as questionnaires are risk-free, an impression that is unintentionally reinforced by the reference to questionnaires in one set of guidelines in the context of "innocuous research".¹⁸ It has been long established that merely knowing that you are taking part in a research study

affects behaviour, irrespective of the intervention.²⁰ There is no reason to think this does not apply in the context of questionnaire-based research.

Whilst the potential harms described above are perhaps less obvious and tangible than those in, for example, clinical drug trials, they still require consideration. Some of the problems associated with the use of questionnaires in primary care research have been highlighted by Jones *et al*, although their emphasis is on interviewer-administered rather than postal questionnaires.¹³ The opportunity for post-interview debriefing for patients, for example, is limited in a postal survey. In the case described, problems were identified in only a very small number of patients but the capacity for harm exists in all similar studies. The conclusions to be drawn from the experience of this case are twofold. The first is the need for robust consent procedures that anticipate and confront the possible harms arising for all participants: clinical subjects, clinical practitioners, and investigating researchers. The second is that the harms discussed above arise from conducting a questionnaire-based study. This demonstrates that simply asking questions is as active an intervention as the administration of a physical treatment, albeit with potential to give rise to psychological consequences (for example, increased anxiety) of a different nature.

The question of whether to intervene in individual cases was raised earlier in the paper. We feel that the study aim should be overridden where necessary, such as would routinely happen in a double blind randomised controlled trial (RCT) where "unblinding" occurs in response to a suspected adverse event. In studies offering anonymity for respondents, opportunities to intervene are limited and the onus is on the respondent to seek assistance. The merits of intervention have been raised in relation to screening surveys where "case-finding" was a primary analytic goal.^{21,22} In contrast, intervention in the current study is considered only following direct requests from patients. The argument for intervention appears more compelling under such circumstances. Whilst the patient may need to initiate such intervention, she may be alerted to this possibility during the consent procedure. This may involve a named contact and clarity about what the patient could expect from her inquiry.

If the method of data collection can be considered an intervention with associated risks, it is reasonable to suppose that the patient also derives personal benefit from participation, over and above the general benefit of the study in contributing to knowledge. Whilst not an aim of the present study, the high response rate, and free-text comments indicate this may be the case. In conclusion, researchers should not be deterred from the important task of investigating health-related behavioural or psychological phenomena. Rather, with the above safeguards in place they can be more confident of minimising some of the deleterious impact for their research participants.

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